doi: 10.1002/ams2.213

Case Report

Emergent transcatheter arterial embolization for norovirusassociated life-threatening ulcer bleeding to achieve successful hemostasis in 2-year-old boy

Wakato Matsuoka,^{1,2} Noriyuki Kaku,^{1,2} Yuichiro Hirata,^{1,2} Sooyoung Lee,^{1,2} Tomohiko Akahoshi,¹ Hiroshi Sugimori,¹ Makoto Hayashida,³ Ryota Souzaki,³ Norihiro Fujita,⁴ Yoshiki Asayama,⁴ Tomoaki Taguchi,³ Hidetoshi Takada² and Yoshihiko Maehara¹

Case: We report a 2-year-old boy with duodenal ulcer with active bleeding that occurred as a result of norovirus gastroenteritis. On admission, the patient presented with shock accompanied with vomiting and melena. Abdominal contrast enhanced computed tomography scan showed signs of duodenal bleeding.

Outcome: He was successfully treated with emergent transcatheter arterial embolization. After the treatment, endoscopic examination revealed duodenal ulcer and the stool norovirus antigen test was found to be positive. The patient recovered completely without any sequelae.

Conclusion: Life-threatening duodenal ulcer bleeding in children can be caused by viral gastroenteritis. When endoscopic therapy is unsuccessful or difficult, in cases of small children, angiographic intervention can be a safe alternative treatment option of gastrointestinal bleeding.

Key words: Norovirus infection, pediatric duodenal ulcer, pediatrics, transcatheter arterial embolization

INTRODUCTION

ASTROINTESTINAL BLEEDING HAS been considered uncommon in children and few cases are reported as a result of gastroenteritis. 1.2 Transcatheter arterial embolization (TAE) is sometimes applied to treat gastrointestinal bleeding in adults, but is rarely applied to small children. We report a 2-year-old boy with duodenal ulcer with active bleeding that occurred as a result of norovirus gastroenteritis. The aim of this case report is to present a rare case of a child successfully treated by TAE. Our report may help clinicians to consider angiographic intervention as a treatment option for pediatric gastrointestinal bleeding.

Corresponding: Noriyuki Kaku, MD, Emergency and Critical Care Center, Kyushu University Hospital, Kyushu University, 3-1-1 Maidashi, Higashi-ku, Fukuoka 812-8582, Japan. E-mail: kakunori@med.kyushu-u.ac.jp

Received 29 Jan, 2016; accepted 5 Apr, 2016; online publication 13 May, 2016

Moreover, we discuss the underlying mechanism of bleeding.

CASE

A 2-YEAR-OLD PREVIOUSLY healthy boy was admitted to the emergency department with sudden onset of massive melena. Four days before his arrival, his younger sister presented with vomiting and watery diarrhea. Norovirus antigen test of her stool was not examined. The next day, our patient showed the same symptoms. He was lethargic even after rehydration therapy at a clinic near his house. He did not complain of abdominal pain at that time. The next day, the patient experienced sudden, massive melena three times and was transported by ambulance to our hospital.

On arrival at the emergency department, he was pale and apathetic with the following vital signs: heart rate, 150 b.p.m.; blood pressure, 72/54 mmHg; respiratory rate 18 breaths/min; and temperature, 37.9°C. The Glasgow Coma Scale was 10 (E4V2M4). Physical examination

¹Emergency and Critical Care Center, Kyushu University Hospital, Fukuoka, Japan, ²Department of Pediatrics, ³Department of Pediatric Surgery, and ⁴Department of Radiology, Graduate School of Medical Sciences, Kyushu University, Fukuoka, Japan

revealed pale conjunctiva and cold extremities. The abdomen was flat without localized tenderness or rebound tenderness, and bowel sounds were hypoactive.

Although he had a history of receiving treatment for bronchial asthma previously, he had no drugs such as corticosteroids or non-steroidal anti-inflammatory drugs before the admission. Fluid bolus therapy was started immediately, but it had no significant effect. Laboratory examination showed severe anemia, with a hemoglobin level of 5.8 g/dL and hematocrit of 17.1%. A red blood cell transfusion was started. In addition, proton pump inhibitor was started i.v. for possible peptic ulcers. Other hematological examination showed a white blood cell count of 14,290/ μ L, a platelet count of 290,000/ μ L, and a C-reactive protein concentration of 1.41 mg/dL.

Abdominal X-ray showed no signs of free air or gut distention. However, abdominal contrast enhanced computed tomography detected a bleeding site at the descending part of the duodenum where the leakage of contrast agents was observed (Fig. 1).

Despite the blood transfusion, the patient did not recover from shock. He was intubated for respiratory assistance. We decided to undertake TAE to stop the bleeding. Angiography showed extravasation at the branch of the anterior superior pancreaticoduodenal artery (Fig. 2). To obtain hemostasis, a 4-Fr Cobra-shaped catheter (Glidecath; Terumo Medical Corporation, Tokyo, Japan) was inserted in the femoral artery. Embolization was carried out with gelatin sponge (Gelpart; Asterals, Tokyo, Japan) and N-butyl-2-cyanoacrylate (Histoacryl; Aesculap, Tuttlingen, Germany) with iodized oil (Lipiodol; Guerbet, Tokyo, Japan) at 1:6

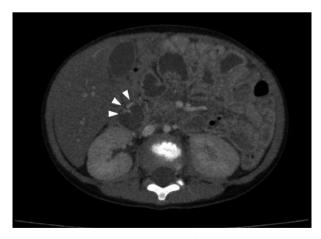


Fig. 1. Computed tomography shows bleeding at the descending part of duodenum (arrowheads) in a 2-year-old boy, resulting from norovirus gastroenteritis.

ratio through a coaxial microcatheter (Carnelian Pixie, 1.8F/2.7F 110 cm; Tokai Medical Products, Kasugai, Japan).

After the treatment, stable vital signs were achieved and hemoglobin levels were maintained within the normal range. One week after TAE, endoscopic examination revealed a duodenal ulcer at the descending part of duodenum (Fig. 3). No sign of vascular malformation, such as pseudoaneurysm of artery, was detected by computed tomography, angiography, or endoscopy. The patient's stool was positive for norovirus antigen. The result of stool culture was negative for pathogenic bacteria. Neither *Helicobacter pylori* nor rotavirus antigen were detected. Serum *H. pylori* immunoglobulin G was also negative. The patient recovered completely and was eventually discharged without any sequelae.

Endoscopic examination showed disappearance of the duodenal ulcer after oral proton pump inhibitor therapy for 2 months.

DISCUSSION

E MERGENT TAE WAS successfully carried out for the treatment of life-threatening intestinal hemorrhage in a 2-year-old boy. In addition, our case was unique because norovirus, one of the common pathogens of gastroenteritis, caused duodenal ulcer accompanied with severe bleeding. Although peptic ulcers had been thought to be uncommon in children, the most common causes of upper gastrointestinal bleeding in children are gastric and duodenal ulcers, esophagitis, gastritis, and varices. The estimated incidence of peptic ulcer bleeding in children ranges from 0.5 to 4.4/100,000 individuals.³ In another study, 5.4% of children who underwent upper gastrointestinal endoscopy because of various gastrointestinal symptoms were diagnosed with peptic ulcer.⁴ These results revealed that the prevalence of peptic ulcers was higher than expected.

Primary ulcers are more common in children aged 10 years and older. The vast majority of primary duodenal ulcers are associated with *H. pylori* infection. In younger children, secondary ulcers occur as a result of external predisposing cause, such as stress or drugs including non-steroidal anti-inflammatory drugs and steroids. Stress-induced ulcers occur in children with respiratory or cardiac distress, sepsis, hypoglycemia, dehydration, accidental injury, or trauma by child abuse. It was reported that the predominant cause of acute upper gastrointestinal tract bleeding in children was duodenal ulcers (75%); *H. pylori* infection was identified in 55% of the patients.⁵

Recently, peptic ulcers have been reported to be associated with gastrointestinal viral infections. Nonetheless, the relation between gastroenteritis and the development of peptic ulcer remains unclear. In our case, norovirus



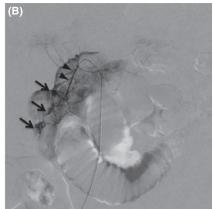


Fig. 2. Catheter angiogram of a 2-year-old boy with duodenal ulcer with active bleeding, resulting from norovirus gastroenteritis. A, Selective angiography of the celiac artery and super selective angiography of the anterior superior pancreaticoduodenal artery (arrowheads). B, Angiography image of the active bleeding. Arrows indicate the contrast extravasation.

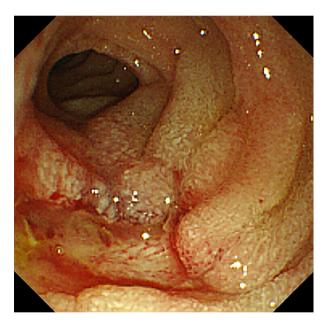


Fig. 3. Endoscopic finding in a 2-year-old boy with duodenal ulcer with active bleeding, a result of norovirus gastroenteritis, 1 week after treatment with transcatheter arterial embolization. An ulcerative lesion of the duodenum is shown.

infection was identified. Although some cases of acute duodenal ulcers with severe melena were accompanied with rotavirus infection, 1,2,6 only three cases were reported in association with norovirus infection. 1,6 Considering the mechanism of bleeding in this case, noroviruses infect mature enterocytes of the duodenal villi, leading to epithelial barrier dysfunction and induction of epithelial apoptosis. Norovirus-induced direct damage to the duodenal wall may be a mechanism of ulcer formation. Although norovirus infection is usually self-limited, necrotizing enterocolitis, 8,9 intestinal perforation, 6 and ulcer such as this case may occur as complications.

Endoscopy is generally effective in treating active bleeding. Upper endoscopy is the diagnostic method of choice for upper gastrointestinal bleeding and permits therapeutic intervention. The safety of diagnostic upper endoscopy has been attested in children, even in infants and neonates. 10,11 However, therapeutic endoscopy is more difficult to apply in younger children. First, deep sedation is required to undertake upper endoscopy in children. Second, smaller caliber endoscopes limit the size of catheters that can be passed through the working channel.¹⁰ Initially, we considered endoscopic treatment, but it was difficult to apply it to a 2-year-old boy in our institute immediately due to the lack of small-sized endoscopes. Finally, the massive bleeding seemed to be difficult to treat endoscopically. Computed tomography showed bleeding from the descending part of duodenum, which meant bleeding from the pancreaticoduodenal artery. Therefore, we chose TAE to stop the massive bleeding from duodenal ulcers and performed it safely without any complications. Surgical treatment was unnecessary.

To the best of our knowledge, this is one of the youngest children to be treated with TAE for massive gastrointestinal bleeding. 1,12 This case report shows that TAE can be a safe alternative to endoscopic treatment and should be considered as the salvage treatment of choice in children with bleeding from duodenal ulcers.

CONFLICT OF INTEREST

NONE.

REFERENCES

- 1 Watanabe E, Tanaka K, Takeda N, Watanabe M. Six cases of life-threatening peptic ulcer bleeding associated with virus infection. J Ped Surg Case Reports. 2014; 2: 269–73.
- 2 Nejihashi N, Sakano T, Ono H, Furue T, Kinoshita Y, Ohta T. An infant with rotavirus infection presenting as a severe acute duodenal ulcer. Pediatr. Int. 2011; 53: 100–1.
- 3 Brown K, Lundborg P, Levinston J, Yang H. Incidence of peptic ulcer bleeding in the US pediatric population. J. Pediatr. Gastroenterol. Nutr. 2012; 54: 733–6.
- 4 Huang SC, Sheu BS, Lee SC, Yang NB, Yang YJ. Etiology and treatment of childhood peptic ulcer disease in Taiwan: a single center 9-year experience. J. Formos. Med. Assoc. 2010; 109: 75–81.
- 5 Houben CH, Chiu PWY, Lau JYW et al. Duodenal ulcers dominate acute upper gastrointestinal tract bleeding in childhood: a 10-year experience from Hong Kong. J Dig Dis. 2008; 9: 199–203.

- 6 Ueda N, Shimotake T, Ohama K. Duodenal perforation associated with norovirus and rotavirus gastroenteritis. Clin Case Rep. 2013; 1: 47–9.
- 7 Karst SM. Pathogenesis of noroviruses, emerging RNA viruses. Viruses 2010; 2: 748–81.
- 8 Pelizzo G, Nakib G, Goruppi I et al. Isolated colon ischemia with norovirus infection in preterm babies: a case series. J Med Case Rep. 2013; 7: 108.
- 9 Turcios-Ruiz RM, Axelrod P, St John K *et al.* Outbreak of necrotizing enterocolitis caused by norovirus in neonatal intensive care unit. J. Pediatr. 2008; 153: 339–44.
- 10 Rahman I, Patel P, Boger P, Rasheed S, Thomson M, Afzal NA. Therapeutic upper gastrointestinal tract endoscopy in paediatric gastroenterology. World J Gastrointest Endosc. 2015; 7: 169–82.
- 11 Colle I, Wilmer A, Le Moine O *et al.* Upper gastrointestinal tract bleeding management: belgian guidelines for adults and children. Acta Gastroenterol. Belg. 2011; 74: 45–66.
- 12 Lee NM, Yun SW, Chae SA, Yoo BH, Cha SJ, Kwak BK. Perforated duodenal ulcer presenting with massive hematochezia in a 30-month-old child. World J. Gastroenterol. 2009; 15: 4853–5.